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Signalling changes to individuals who show resistance to change can reduce challenging behaviour

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Signalling changes to individuals who show resistance to change can reduce challenging behaviour

Abstract

Background & objectives

Several neurodevelopmental disorders are associated with resistance to change and challenging behaviours – including temper outbursts – that ensue following changes to routines, plans or expectations (here, collectively: expectations). Here, a change signalling intervention was tested for proof of concept and potential practical effectiveness.

Methods

Twelve individuals with Prader-Willi syndrome participated in researcher- and caregiver-led pairing of a distinctive visual-verbal signal with subsequent changes to expectations. Specific expectations for a planned subset of five participants were systematically observed in minimally manipulated natural environments. Nine caregivers completed a temper outburst diary during a four week baseline period and a two week signalling evaluation period.

Results

Participants demonstrated consistently less temper outburst behaviour in the systematic observations when changes imposed to expectations were signalled, compared to when changes were not signalled. Four of the nine participants whose caregivers completed the behaviour diary demonstrated reliable reductions in temper outbursts between baseline and signalling evaluation.

Limitations

An active control group for the present initial evaluation of the signalling strategy using evidence from caregiver behaviour diaries was outside the scope of the present pilot study. Thus, findings cannot support the clinical efficacy of the present signalling approach.

Conclusions

Proof of concept evidence that reliable pairing of a distinctive cue with a subsequent change to expectation can reduce associated challenging behaviour is provided. Data provide additional support for the importance of specific practical steps in further evaluations of the change signalling approach.

Key words

preference for routine; tantrum; intellectual disability; autism; stimulus control; resistance to change; Prader-Willi syndrome

1. Introduction

Several neurodevelopmental disorders are associated with a strong preference for predictability, with increased risk of challenging behaviour following changes to routines, expectations or plans (will be referred to collectively here as expectations). For example, resistance to change is more common in individuals with Prader-Willi, fragile X, Smith Magenis, and Lowe syndromes compared to multiple neurodevelopmental disorder comparison groups (Moss, Oliver, Arron, Burbidge & Berg, 2009). The increased risk of challenging behaviour following changes to expectations has been demonstrated in research with individuals with Prader-Willi and fragile X syndromes (Tunnicliffe, Woodcock, Oliver, Bull & Penhallow, 2014; Woodcock, Oliver & Humphreys 2009a) and is reported anecdotally by families with these other genetic syndromes. Further, changes to expectations have been demonstrated as a common trigger of challenging behaviour in individuals with an intellectual disabilities of mixed aetiologies (Furniss & Biswas, 2012), and in individuals with autism spectrum disorder (Gomot & Wicker, 2012).

Prader-Willi syndrome[1], one such disorder, has been estimated to have a population prevalence of at least 1:52,000 in the UK, although the actual rate is likely to be somewhat higher (Whittington et al., 2001). The disorder is associated with mild to moderate intellectual disability, with an average IQ of around 60 (Whittington et al., 2004). PWS is caused by a mutation affecting the paternally derived q11-q13 region of chromosome 15. Most prevalence estimates for clinically elevated preference for predictability are upwards of 60% of individuals with the syndrome (Holland et al., 2003; Moss et al., 2009). The profile of challenging behaviour precipitated by changes in people with PWS most commonly takes the

[1] Prader-Willi syndrome is abbreviated as PWS

form of temper outbursts, which are shown by at least 80% of individuals and are an important priority for intervention (Holland et al., 2003; Tunnicliffe et al., 2014; Woodcock et al., 2009a).

The objective of the present study was to develop and pilot a caregiver led behavioural intervention to decrease the frequency and severity of temper outbursts triggered by changes to routines, expectations and plans (referred to collectively as changes to expectations) shown by individuals with PWS. Given the expression of similar resistance to change behaviour in individuals with other neurodevelopmental disorders, this work will provide an important foundation for the application and evaluation of such an intervention approach on a larger scale.

When individuals with PWS are exposed to changes to expectations across different settings but with the consistent presence of a particular stimulus (e.g. a specific person), there can be a reduction in the frequency of low level challenging behaviours precipitated by such changes over successive experimental observations (Woodcock, Oliver & Humphreys, 2011). A possible explanation for this effect is that via repeated pairing with changes to expectations, the stimulus comes to reliably predict the occurrence of changes through associative learning, and that this increased predictability makes the change less aversive.

This explanation draws support from the specific cognitive difficulty in task switching, which appears to be linked to the preference for predictability observed in individuals with PWS via the demand that changes to expectations place on such switching abilities (and may be relevant for the corresponding preference observed in certain other neurodevelopmental disorders, such as fragile X syndrome and autism spectrum disorder; D'Cruz et al., 2013; Lopez et al., 2005; Woodcock, Oliver & Humphreys, 2009b). Neurocognitive paradigms have demonstrated that task switching can be facilitated by presentation of external stimuli

that indicate a particular task, and also by increasing the time available to prepare for a switch once knowledge of its pending occurrence has been ascertained (e.g. Monsell, 2003). Thus, the presence of a stimulus that reliably predicts the occurrence of a change to expectation may reduce the demand on the deficient cognitive process linked to resistance to change.

The reliable pairing of a distinctive cue with an event or stimulus that demonstrates a known relationship with an individual's behaviour has been exploited in stimulus control procedures, which form an important part of several behaviour intervention approaches that have demonstrated utility with individuals with neurodevelopmental disorders (Shahan & Chase, 2002). Distinctive cues have been paired with the non-availability of reinforcing contingencies for challenging behaviour so that challenging behaviour is reduced in the presence of the cue (e.g. Cammilleri, Tiger & Hanley, 2008; Heald, Allen, Villa & Oliver, 2013; Kuhn, Chirighin & Zelenka, 2010). In addition, distinctive cues have been paired with current or upcoming aversive stimuli (a verbal reprimand, the removal of a preferred item/activity) so that eventually challenging behaviour is reduced in the presence of the cue alone (Maglieri et al., 2000).

The reductions in change-triggered challenging behaviours observed in individuals with PWS over successive changes when a particular person is present (Woodcock et al., 2011) suggest that pairing a cue with the reliable occurrence of changes may constitute an effective intervention strategy. To the best of our knowledge, stimulus control procedures have not been previously applied in this context in any population (i.e. using a cue to signal impending changes – regardless of their nature – when change per se has been identified as a key antecedent for challenging behaviour). However, video modelling has been applied, particularly with individuals with autism spectrum disorder, aiming to increase the ease of individuals' transitions between tasks (e.g. Schreibman, Whalen & Stahmer, 2000), and it has

been suggested that such modelling results in the ascertainment of stimulus control by the video over transitioning behaviour (Nikopoulos, Canavan & Nikopoulou-Symrni, 2009). Thus, such video modelling approaches appear to increase the predictability of impending events in an activity specific way. Similarly, visual activity schedules have been widely employed in transition settings with individuals with autism spectrum disorders and intellectual disabilities. Frequently such approaches have aimed to increase transitioning behaviour (i.e. have a primary goal and measured outcome of increasing adaptive functioning, not of reducing challenging behaviour) and current cumulative evidence supports the efficacy of visual scheduling in achieving this objective (Knight, Sartini & Spriggs, 2015). However, there is also evidence demonstrating that use of visual schedules can decrease challenging behaviour linked to transitions (e.g. Mesibov, Browder & Kirkland, 2002; Tullis, Cannella-Malone & Payne, 2015). Whilst visual activity scheduling may potentially reduce the number of changes to expectations that individuals are exposed to (because the sequence of events described in the schedule is adhered to), the content of the schedule may also increase the predictability of impending events in a task specific way, similarly to the video modelling approach.

In the present study, a caregiver led intervention strategy for reducing temper outbursts triggered by changes to expectations in individuals with PWS was developed, implemented and evaluated in a proof of concept study (such studies have been highlighted as an essential step in intervention development; Craig et al., 2008). A stimulus control approach was used to establish a distinctive signal, which would reliably predict the occurrence of a change to expectation. The novelty of the present approach results from its independence from the nature of the impending changes in an individual's environment. Presently employed and previously evaluated intervention approaches that attempt to increase

the predictability of individuals' environments do so in an event dependent manner (i.e. by increasing information available on impending activities as in the video modelling and activity scheduling procedures described above). The present approach however, requires no specific information on forthcoming activities and thus has the potential to be more resource efficient and easier to implement than existing approaches.

We hypothesized that stimulus control over temper outburst behaviours would be demonstrated such that these behaviours would be lower in frequency following a change to expectation that was signalled, compared to a corresponding change that had not been signalled. In addition, relative to a baseline period preceding application of the signalling procedure by caregivers, the overall number of temper outbursts following changes to expectations would decrease.

2. Material and methods

2.1. Participants

Twelve individuals with PWS were recruited via the Prader-Willi Syndrome Association in the UK – a support group for families – and from a group of specialist UK residential homes. In line with the demographic makeup of the support association, all participants were white British and of middle to high socioeconomic status. Caregivers were interviewed via telephone on the context of the temper outbursts that they observe (see *Appendix A* for the interview schedule). Inclusion criteria specified that individuals must display frequent temper outbursts (at least 2-3 per week on average); and that changes to expectations should constitute a commonly occurring antecedent event for these outbursts. Notably, the present intervention approach was specifically designed to be appropriate for any individual demonstrating temper outbursts following changes to expectations, and the present

study attempts only to draw inferences about such individuals (these behaviours are commonly but not universally shown by individuals with PWS). Thus, confirmed diagnosis of PWS did not comprise an inclusion criterion.

All families had previously participated in the studies described in (Bull, Oliver, Tunnicliffe & Woodcock, 2015; and Bull, Oliver, Callaghan & Woodcock, 2015), which included the development of the presently applied behaviour diary, and table top games used here in the *researcher led teaching* (below). The telephone interview was conducted at the beginning of the participation timeline across all studies (which ran over up to 24 months). Ethical approval for the study was obtained from the University of Birmingham Ethics Review Committee. All participants aged sixteen years or older gave written informed consent to take part; children under 16 years provided written assent and their parents provided written consent. Demographic information for participants is described in *Table 1*.

[*Table 1*]

2.2. Signal

A cue card (*Figure 1*) was developed and checked for novelty with families. The signal comprised presentation of the card along with the verbal phrase "this picture means something different is going to happen".

[*Figure 1*]

2.3. Overall procedure

2.3.1. Research activities involved

Figure 2 illustrates the research activities involved in overall experimental procedure, details of which follow in *Sub-procedure* and *Measures* sections. Research activities included

1. Researcher-led teaching of the signal (Section 2.4), 2. Caregiver-led extended teaching of the signal (Section 2.5), 3. Natural evaluation (of the signalling; Section 2.6) and 4. Mechanistic evaluation (of the signalling; Section 2.7).

[*Figure 2*]

2.3.2. Participants' involvement in research activities

All caregivers took part in the baseline recording (4 weeks in duration) of temper outbursts in participants' natural environments using the behaviour diary (see *Measures*) before initiation of other research activities. Following researcher-led teaching of the signal, three participants withdrew from the study due to changing demands on the families. Thus, 9 participants took part in the caregiver-led extended teaching of the signal and the natural evaluation. For the mechanistic evaluation, given the associated heavier demands on participants and researchers, a sub-sample of five participants was planned. Participants were invited to take part in the mechanistic evaluation based on their availability at times that best fitted with the research schedule (for example, participants who lived far from the research base and/or who had a particularly full schedule of activities that limited their free time, agreed that this part of the study would not be appropriate for them). Importantly, selection criteria were entirely pragmatic and no analysis of other data collected had taken place at the time of selection. To obtain the sub-sample of five, six families were invited to participate but one family was not willing to do so because of concerns about possible upset for the participant.

Of note, all research activities included both children/adolescents and adults (see *Table 1*). However, the only female who participated withdrew from the study following researcher-led teaching of the signal.

2.4. Researcher led teaching of signal sub-procedure

2.4.1. Table-top games with participants

Participants had previously engaged with two table-top games – for 40 and 80 minutes respectively – during which routines were established (see Bull, Oliver, Callaghan et al, 2015). For example, one game involved specific action with cards, dice and counters; and included a routine of separating cards into two piles after each player's turn.

Each game was played for thirty minutes during alternating five minute conditions in which routines were followed without change, or changes were imposed to routines but these were preceded by presentation of the signal. Over the hour, the researcher checked five times that the participant could answer affirmatively that the picture meant that something different would happen. One participant did not wish to engage with the games, so eleven participants took part in this procedure.

Following this, two test sessions were conducted (one in the context of each game), each comprising three, five minute counterbalanced conditions. Critically, the conditions allowed comparison of participants' responses to changes that were preceded with the signal, compared to changes that were not signalled. However measurements of behaviour and physiological arousal did not provide evidence that the signal to change association held by participants was already strong enough to effect behavioural change (see *Appendix B*). Thus, further training in participants' natural environments was deemed necessary.

The table top games with participants ended with four alternating five minute long sessions (in the context of the game previously played for less time) during which either no changes were imposed; or regular changes were preceded by presentation of the signal.

2.4.2. Demonstration with caregivers

Written instructions were given to caregivers on how to use the signal (see *Appendix C*) and a researcher ran through these verbally with examples. Emphasis was placed on the importance of reliable signal to change pairing. Caregivers maintained their typical pattern of responding in the context of changes except for the added presentation of the signal. Following explanation of the instructions, the researcher observed the caregiver explaining the signal card to the participant and asked whether the participant knew what the card meant. The researcher then also observed the signal being used with the participant once (in the context of a small imposed change (see *Section 2.5* for the definition of a small change), not expected to trigger any upset or temper outburst behaviour), and discussed this with the caregiver along with any issues the caregiver foresaw related to future use.

For the children (n=4, *Table 1*) who were regularly exposed to both home and school settings, the demonstration was carried out with a caregiver from both settings (n=2), or teachers received written instructions and opportunity to ask questions via telephone (n=2, where research presence at school may have caused upset).

2.5. Caregiver-led extended teaching of the signal sub-procedure

It was important to allow the signal to be learned in participants' natural environments, whilst avoiding scenarios in which it may become associated with aversive events. Thus, an individualised, three level hierarchy of changes to expectations, was developed with caregivers. The lowest level comprised changes expected to be perceived positively by participants (e.g. to a preferred event); followed by "small" changes, expected to trigger lower level upset and temper outburst behaviours; finally, "big" changes were those expected to trigger severe temper outbursts.

Over one week, using the hierarchy and as described in the signalling information sheet (*Appendix C*), caregivers were asked to impose deliberate changes – which would be signalled – once every two days, from those in the lowest level of the hierarchy (i.e. positively perceived changes). In addition, although caregivers were asked to use the signal preceding naturally occurring changes, they were asked specifically *not* to use the signal if any changes from the highest level of the hierarchy occurred (i.e. would usually trigger the most severe temper outbursts). During the following week, caregivers continued with the imposition of changes from the lowest level of the hierarchy every two days; but also signalled all possible naturally occurring changes. In addition, whenever use of the signal was followed by adaptive (non-temper outburst) behaviours by a participant, caregivers were asked to provide social praise. Thus, the caregiver-led extended teaching of the signal aimed to combine positive reinforcement with presentation of the signal during participants' initial learning periods.

2.6. Natural evaluation of signalling sub-procedure

Caregivers were asked to make full use of the signal by using it to precede all changes of which they had some advance warning. No deliberate changes were imposed by caregivers during this period. Caregivers completed the behaviour diary (see *Measures*) during this period to record any temper outbursts that occurred but researchers were not present and the only manipulation of participants' environments was the use of the signal by caregivers when appropriate. The primary purpose of this natural evaluation period was to assess the feasibility of use of the signal by caregivers in participants' natural environments; and the feasibility of caregivers recording of temper outbursts using a behaviour diary during their use of the signalling strategy.

2.7. Mechanistic evaluation of signalling sub-procedure

The mechanistic evaluation comprised the primary test of proof of concept of the signalling intervention strategy by systematically comparing participants' responses to specific routines or expectations when these were either followed as expected (no temper outburst behaviour would be predicted); changed unexpectedly (temper outburst behaviour would be predicted); or changed following presentation of the signal (less temper outburst behaviour compared to when the change was unexpected would support proof of concept). Thus, three types of observations (comprising an observation set) were made of participants in their natural environments. Observations were conducted in the presence of a researcher, who video recorded the procedure (*see Measures 2.7.2*). A routine was observed without any change, an equivalent routine was observed with a change imposed *without the signal*, and finally an equivalent routine was observed with a *signalled* change. Changes were imposed by caregivers. Routines observed were selected from the middle of participants' hierarchies (i.e. expected to elicit some, but not severe, temper outburst behaviours: *Table 1*) to minimise potential distress and allow for repeated observations. The order of the observations within a set was variable across participants, based on restrictions imposed by the participants' environments. Thus, observation order was approximately counter-balanced across participants within observation sets. Up to three observation sets were conducted with each participant (as could be accommodated by families). Observation sets were made on different days during the natural evaluation period as was convenient for families. Importantly, caregivers were not asked to make behaviour diary entries corresponding to the time when the researcher was present for the mechanistic evaluation and the two weeks of diary recording during the natural observation period excludes the times when the researcher was present to conduct the mechanistic evaluation.

2.7. Measures

2.7.1. Informant report behaviour diary

Caregivers completed a behaviour diary documenting temper outbursts during the entire study period (*Figure 2*), including during a four week baseline period prior to initiation of the other research activities. Entries included the date, time, duration and an intensity rating (ten-point scale). In addition, component behaviours, triggers, and after events were recorded (see *Appendix E*). Importantly, this approach provided the means to evaluate signalling strategy mediated changes in temper outbursts triggered by changes to expectations. In previous research with the present participants, recordings of heart rate and activity level have indicated that physiological arousal is consistently elevated during periods when caregivers report outburst occurrences. In addition, high concurrence between outburst nature as described in diaries and informant report structured interviews was demonstrated (Bull, Oliver, Tunnicliffe et al., 2015). Importantly, this previous examination of the validity of the behaviour diaries did not allow investigation of the validity of the 10-point intensity ratings. Further, not all participants completed the intensity rating for each outburst recorded in the diary. Thus, data on reports of outburst intensity are not reported further.

2.7.2. Behaviour observation

Participants were video recorded during the mechanistic evaluation sub-procedure. Observed behaviours were operationally defined and two researchers independently coded at least 25% of the footage. Inter-rater reliability was ascertained and any behaviour categories with low reliability were collapsed until a coding system of high reliability was ascertained (*Appendix D*). Temper outburst behaviours coded were ignoring requests, arguing, crying,

verbal aggression (Kappa 0.87-0.96), questioning (Kappa: 0.62) and picking nose (Matthew only; Kappa: 1.0).

2.7.3. Open-ended anecdotal reports

Caregivers were contacted each week during the intervention to check progress and to gather anecdotal reports about the signalling strategy. The researcher asked "how is the cueing strategy going?" as a non-leading question designed to minimise demand characteristics in caregivers' reports.

3. Results

3.1. Mechanistic evaluation of signalling

For James, to minimize disruption to daily activities, one observation from the second set was conducted on the same day as the first observation set. For Matthew, the first observation conducted during the first set involved a signalled change to routine that elicited substantial temper outburst behaviour (*Table 1*). It was deemed inappropriate by caregivers and the researcher to continue with the other observations in that set, as caregivers expected that more temper outburst behaviour may ensue than was reasonable for research purposes.

The percentage of each observation during which participants' temper outburst behaviours were observed is illustrated in *Figure 3*. Importantly, temper outburst behaviours were consistently reduced during observations in which changes were signalled, compared to corresponding observations in which changes were not signalled.

[*Figure 3*]

3.2. Naturalistic evaluation of signalling

Reported temper outbursts triggered by changes to expectations were extracted from participating families' behaviour diaries during the four week baseline (prior to initiation of the other research activities, including signalling training) and two week naturalistic evaluation period.

To ascertain an estimate with maximum possible stability of the rate of change triggered temper outbursts during a two week baseline period for each participant (to correspond with the naturalistic signalling evaluation duration), the number of these outbursts shown during each week was added to those shown during each other week to give six rates of change triggered temper outbursts corresponding to different two week periods. Non-parametric bootstrapping was then applied with 5000 iterations to create a distribution of mean change triggered temper outbursts during a two week baseline period. The mean of this distribution was taken as the estimated mean change triggered temper outbursts during a two-week baseline and the 5^{th} and 95^{th} percentiles of this distribution comprised the 95% confidence intervals around the mean. Such non-parametric bootstrapping is not restricted by the assumption of a population distribution of a particular shape (Kline, 2013). However, it must be noted that the method is typically applied to larger samples of independent observations and thus its application to within participant observations has not been widely tested. Nevertheless, the confidence intervals calculated produce an estimate of what would constitute reliable change, which would otherwise not be possible since the temper outburst diaries have only recently been developed. Before applying the bootstrapping procedure, weekly records of temper outbursts were examined (*Appendix F*). Consistent trends over this period were not evident but substantial week to week variability supported the importance of considering the present estimates of reliable change.

Five of the nine participants showed a reduction in the number of diary reported change triggered temper outbursts between baseline and naturalistic signalling evaluation (*Figure 4*). Four of these five demonstrated reductions that were beyond the lower bound of the corresponding 95% confidence intervals, suggesting that for one participant (Daniel) the observed change may not have been reliable. One individual (Matthew) showed no difference in the number of change triggered temper outbursts demonstrated at baseline and during naturalistic signalling evaluation. Peter and James demonstrated an increase in the number of change triggered temper outbursts between baseline and naturalistic signalling evaluation, which went beyond the upper bound of the corresponding 95% confidence intervals. Across all participants, the frequency of change triggered temper outbursts reported during the whole behaviour diary period was low, with Daniel demonstrating the highest rate of these outbursts; three and a half during the two week baseline period.

[*Figure 4*]

3.3. Follow up

The caregivers of Daniel and Alfred, who continued to implement the signalling strategy following the end of the naturalistic evaluation period, completed more temper outburst diary entries.

For Daniel, these reports related to an eight week period immediately following the naturalistic evaluation and indicated an average of four change triggered temper outbursts for each two week period of the first four weeks, and three relevant temper outbursts for each two week period of the second four weeks. These data further support the suggestion that the observed reduction in number of change triggered temper outbursts between baseline and naturalistic evaluation period for Daniel may not have been reliable.

For Alfred, the reports related to a four week period that began 37 days after the naturalistic evaluation period ended. During this time, one change triggered temper outburst was demonstrated during each two week period. This remains lower than the baseline rate of temper outbursts reported for Alfred but not below the lower bound of the corresponding 95% confidence interval of that baseline rate.

3.4. Open ended anecdotal reports

Anecdotal reports received (see *Appendix F*) indicated generally positive perceptions of the signalling strategy by caregivers. Interestingly, two participants' caregivers (Charles, Bob) stopped using the original signal comprising the card with the standard verbal phrase and instead started using the verbal phrase "change" in otherwise the same way as the signal. Several reports indicated that the signalling strategy was more often successful with certain changes compared to others and that changes with food routines or during contexts of increased stress for participants, remained difficult to manage for some participants whether caregivers signalled the change or not.

4. Discussion

A change signalling strategy was implemented by the caregivers of participants recruited based on the occurrence of frequent temper outbursts, with some of those outbursts being reported following unexpected changes to participants' expectations. All participants were believed to have Prader-Willi syndrome, however a confirmed genetic diagnosis of the syndrome did not constitute an inclusion criterion. The signalling strategy was implemented following researcher- and caregiver-led teaching of the signal-to-change association in a

controlled and natural environment respectively. Following participants' engagement with the teaching procedures, all individuals who were systematically observed in their natural environments (mechanistic evaluation) showed consistently reduced temper outburst behaviour following a change to expectation that was signalled, compared to a corresponding change that was not signalled. These observations support our hypothesis and provide proof of concept evidence that it is possible to decrease individuals' change triggered temper outburst behaviours by preceding changes with a distinctive cue that only occurs when a change is about to happen (thus, increasing the predictability of such changes). Furthermore, results from temper outburst diaries (naturalistic evaluation) – kept by caregivers before and during implementation of the signalling strategy – indicated that signalling may have mediated reductions in change triggered temper outbursts for some participants, even over the short period that was included in the present pilot evaluation. Anecdotal reports revealed important considerations for future development of the change signalling strategy.

When considering the implications of the present results, it is important to expand on the difference between the mechanistic and naturalistic evaluations in terms of the evidence each provides with respect to the potential effectiveness of wider scale implementation of the change signalling strategy. The proof of concept evidence provided by the mechanistic evaluation was ascertained in the context of relatively low level temper outburst behaviours. Such temper outburst behaviours have been manipulated in previous research to demonstrate relationships between temper outbursts and environmental contingencies (Bull, Oliver, Callaghan et al., 2015; Oliver et al., 2009; Woodcock et al., 2011). It has also been demonstrated that manipulation of such precursor behaviours in an intervention setting can produce beneficial effects on the corresponding more challenging behaviours (Langdon & Carr, 2008). Thus, the mechanistic evaluation results are those that are critical in supporting

the case for further evaluation of the signalling strategy. The behaviour diary data ascertained during the naturalistic evaluation on the other hand, relates to full temper outbursts.

Results for Peter and James nicely illustrate the contrast between mechanistic and naturalistic evaluation results. These participants showed reliable reductions in low level temper outburst behaviours when a signal preceded a particular change to expectation relative to when no such signal was provided. However, both participants demonstrated an (albeit small) increase in the number of full change triggered outbursts reported in the diaries between baseline and signalling evaluation phases (and were indeed the only two participants to demonstrate such increases). It is possible that uncontrolled environmental factors led to the occurrence of more changes to expectations that these participants found particularly difficult during the naturalistic signalling evaluation relative to the baseline period. This explanation derives some support from the anecdotal reports about Peter, which refer to changes being more difficult when stressors build up in his environment. On the other hand, it is also possible that the signalling strategy would be effective only for lower level challenging behaviours in these participants. Supporting this possibility, an important interaction between resistance to change and the occurrence of a preferred event has been demonstrated in previous research examining the efficacy of intervention approaches aiming to increase environmental predictability (Waters, Lerman & Hovanetz, 2009). Further research evaluating the present signalling strategy, which tracks occurrence of changes to expectations during baseline and evaluation periods (whatever the result of those changes), will be important to distinguish between these possibilities.

The limitations of the naturalistic signalling evaluation approach are also important to bear in mind. Rates of change triggered temper outbursts during the two week baseline were low. Whilst the present analyses attempted to compensate for the low rates of temper

outbursts observed during baseline, an approximate estimate of the reliability of the rate of relevant temper outbursts could be provided only. These low rates were evident despite inclusion criteria specifying at least 2-3 outbursts per week. In the present study – to work within the level of resources available – inclusion criteria were checked shortly after initial recruitment, several months before initiation of the baseline period reported. One factor contributing to the low baseline rates of change triggered temper outbursts reported here is therefore a possible maturational trend (reduction over time) in the frequency of these behaviours. Structured interviews with the present caregivers have indicated stability in participants' temper outbursts over a six month period (Tunnicliffe et al., 2014), suggesting that such maturational trends are unlikely to have been the only factor contributing to the low baseline rates of change triggered temper outbursts. Nevertheless, the low rates of baseline temper outbursts observed here, combined with the possibility of maturational trends in outbursts being present, point towards the importance of employing long baseline periods when conducting future evaluations of interventions for temper outbursts. Given the demands on participating families, particularly during intervention baseline, this evidence is crucial for supporting future optimal evaluation designs.

An additional factor that may have contributed to the low rates of baseline change triggered temper outbursts was the presently employed inclusion criteria. Here, initial recruitment materials specified that the study required participants evidencing difficulties with change, who often show temper outbursts following changes. Thus, in structured interviews with the present participants, change to routines/expectations was the most common trigger for temper outbursts (Tunnicliffe et al., 2014). However, the present inclusion criteria required only that participants showed a minimum of 2-3 outbursts per week. The proportion of those outbursts that should be preceded by changes was not specified. Therefore, for the

future development of intervention approaches based on the presently applied signalling, it will be important to carefully examine possible differences in the observed efficacy of the signalling approach for individuals who show different initial rates of change triggered temper outbursts. With the development of a valid measure for ascertaining the frequency of a potential participants' change triggered outbursts at the point of assessment of their eligibility for entrance into the intervention, such knowledge of individual characteristics associated with greater observed benefit of signalling participation would provide the basis for tools capable of informing an individual family whether the approach is likely to be beneficial for them.

The final factor important to highlight here, which may have contributed to the low baseline rates of change triggered temper outbursts, is the highly intense nature of temper outbursts (e.g. Potegal, 2003; Wakschlag et al., 2012). Such intensity may result in a bias towards the behaviour being perceived by caregivers as more frequent than objectively measured rates. This factor highlights the value of continuing to develop the presently employed behaviour diary method for monitoring temper outbursts, which is less subject to such subjective biases than informant report interviews or questionnaires. Further, it is possible that moving forward, a behaviour diary approach to initial assessment of whether a signalling intervention approach is likely to be effective for an individual may be beneficial.

Anecdotal reports of the adoption of a different signal by two out of the nine participating families suggest that in a wider scale implementation of the intervention it would be important to integrate flexibility into the signal. In addition, the reports of increased difficulties with certain types of changes – for example when there are increased concurrent stressors or changes around food – support the caregiver-led extension of signal teaching approach that was adopted in the present study, which made the distinction between different

types of changes during initial signal acquisition. It seems likely that in future studies, lengthening the teaching in participants' natural environments may be beneficial.

Although the primary objective of the present study was to provide the proof of concept evidence necessary to support future change signalling evaluation studies, the limitation relating to the lack of a control intervention in the evaluation of the naturalistic signalling must be highlighted. Major placebo effects have been reported on caregiver perceptions of children's temper outbursts (Whalley & Hyland, 2013). Future evaluation studies should therefore employ an appropriate comparison intervention, particularly if an informant report measure will comprise a primary outcome.

Finally, the present procedure did not allow the results of a pure signalling (stimulus control) approach to be separated from the effects of positive reinforcement (included deliberately in signal teaching). This procedure was adopted based on the assumption that reinforcement would facilitate learning. However, future research is needed to examine the potentially synergistic (or even primary) contribution of such reinforcement to the effects of signalling changes.

5. Conclusions

The results of the present study describe critical steps in the development of a novel treatment approach for challenging behaviour associated with resistance to change shown by individuals with neurodevelopmental disorders. Although the present findings cannot be taken to support the clinical efficacy of the treatment approach (since inclusion of an active control group was outside the scope of the present study), the findings provide proof of concept evidence that reliable pairing of a distinctive signal with forthcoming changes, is

capable of reducing the extent of individuals' challenging behavioural responses to change. Further, practical information on the importance of extended baseline behaviour measurement periods for future evaluations, and on how best to implement the treatment approach, were ascertained.

6. Acknowledgements

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Table 1 Participant demographics and observations for those who took part in the mechanistic evaluation sub-procedure

Participant[2]	Research activities	Age (y:m)	Gender	Genetic subtype	VABS[3]	Observation setting	Caregiver involved	Routine observed	Expectation changed
Scott	All teaching of signal; Natural evaluation; Mechanistic evaluation	33:4	Male	UPD	25	Care home	Support worker	Snack time	Type of food
Bob		47:10	Male	NA	62	Care home	Support worker	Daily planned activity	Nature of activity
Matthew		16:4	Male	NA	56	Home	Mother	Food eaten	Type of food
						School	Support worker	Snack time	Time (delayed)
Peter		45:8	Male	UPD	61	Care home	Support worker	Washing clothes	Clothes to be washed
James		10:3	Male	UPD	95	Outdoors	Mother	Dog walk	Path taken
John	All teaching of signal; Natural evaluation	39:7	Male	NA	57				
Daniel		10:9	Male	UPD	61				
Alfred		43:1	Male	del + trans	40				
Charles		9:5	Male	NA	76				
Flora[4]	Researcher-led teaching of signal	24:7	Female	del	52				
Alex		11:8	Male	UPD	70				
Richard		9:7	Male	UPD	79				

[2] Pseudonyms are used to protect confidentiality, all participants are male

[3] Vineland Adaptive Behavior Scales (VABS; Sparrow, Cicchetti & Balla, 2005) total composite scores can be converted to the adaptive level described as low, moderately low, adequate, moderately high or high. All means fell in the low range (<70), with the range of scores spanning low to adequate. Genetic diagnoses comprised a paternal deletion in the q11-q13 region of Chromosome 15 (del); a maternal uniparental disomy of Chromosome 15 (UPD); a translocation on Chromosome 15 (trans) or caregivers did not have access to a genetic diagnosis (NA).

[4] Participants described in grey typeface are those that took part only in the researcher-led teaching of the signal

Figure 1: the signal card that was laminated and presented to participants before changes along with the verbal phrase “change”. Height was 91mm; width was 58mm

Figure 2: overall experimental procedure. ME refers to the mechanistic evaluation sub-procedure. Parts of the procedure when a researcher was present are indicated (RP).

Figure 3: The percentage of mechanistic evaluation observations when temper outburst behaviours were shown is plotted relative to the type of observation (whether no change was made or whether a change was presented with or without the signal). Bar clusters indicate observation sets for each participant, ordered chronologically. Individual observations in each set took place in varying orders to best fit into participants’ daily activities and so were approximately counter-balanced across participants (illustrated here in a fixed order for ease of visualisation).

Figure 4: Number of temper outbursts reported in the behaviour diary for each participant during the natural evaluation of signalling (2 weeks) relative to the bootstrapped mean number of temper outbursts reported during a two week baseline period

Appendices

Appendix A. **Semi-structured Interview Schedule for recruitment and creating the behaviour diary (Telephone)**

Temper outburst/tantrum –

Highly emotional response. Period of crying, screaming, angry ranting, shouting, stamping feet, or kicking. Can last for a prolonged period of time.

1. Does _____ ever display temper outburst behaviour?

2. If so, what behaviours does _____ show during a typical episode?

3. How often do the temper outbursts occur?

4. Think about the last time a temper outburst occurred, what seemed to trigger the behaviour in this example?

5. In the example you thought about, how did you respond to the temper outburst?

6. Roughly, how many times does the trigger you mentioned actually result in a temper outburst?

7. Are there times when this particular trigger does not actually trigger a temper outburst?

8. Roughly, how many times after a temper outburst would you respond in the way that you mentioned in the example?

9. What behaviours does _____ typically show after an outburst?

10. Think of other examples in which a temper outburst occurred, what seemed to trigger this and how did you respond?

11. How long roughly do the temper outbursts last for?

Review

12. Finally go over behaviours, antecedents and consequences listed.
    Anymore behaviours, antecedents, consequences?

Can these antecedents and consequences be categorised? Do this with parent/carer.

Appendix B. Researcher-led teaching of signal sub-procedure - table top games with participants

Test session method

Behavioural observations

Participants were filmed using a video camera. Behaviours of interest were any temper outburst related behaviours and these were coded using the computer package ObsWin 3.2 (Martin, Oliver & Hall, 2000) that allows for the real time coding of behaviour. Behaviours coded were all behaviours that parents or carers had identified during the semi-structured interviews. Behaviours of interest were operationally defined with some definitions from Oliver et al. (2009) being useful. Inter-rater reliability was established for 25% of each participant's data. Kappa values of above .6 were established for all behaviours indicating good inter-rater reliability. See *Table B1* for information on reliability.

Table B1 Behaviours coded during observations of participants during test sessions in researcher-led signal teaching procedure

Behaviour Coded	Operational Definition	Inter-rater reliability: Kappa
Questioning	The participant asks the researcher a question related to the game. These could be about the rules/materials/turns.	0.82
Ignoring Requests	The participant does not respond to a verbal request made by the researcher or the participant starts to verbalise about something unrelated to the request. This should be coded until a further verbal response from the researcher (either a further request or a verbalisation about something unrelated to the request) or the participant stops ignoring and initiates a response.	0.60
Arguing	The participant makes verbalisations in the form of statements of disagreement, giving order or making demands, taken from Oliver et al. (2009).	0.68
Physical Aggression	The participant responds with a deliberate act towards researcher or object involving contact that could cause harm or damage. This should also include any missed attempts at physical aggression where no contact is made.	1

Physiological recordings

Participants wore a heart rate monitor (Polar RS400; to measure heart rate) and an Actiwatch that measures activity whilst playing the games. Heart rate information was collected as an indicator of level of physiological arousal (different emotions increase heart rate (Schwartz et al., 1981, Ekman et al., 1983, Sinha, et al., 1992, Rainville et al., 2006 and Fernandez et al., 2012), whilst the activity data were used to exclude the potentially confounding influence of physical activity level on heart rate as an indicator of arousal. Increases in heart rate can be caused by physical exercise (Iellamo, 2001).

Heart rate was recorded in beats per minute (bpm) and recorded every one second. Activity was recorded as an activity count. The Actiwatch has an accelerometer, when movement is detected it produces an electric current and any change in voltage is measured as an activity count. Activity counts were recorded in epochs every ten seconds.

Test session results

Mean percentage of time during which temper outburst behaviour was presented during test conditions including no changes, un-signalled changes and signalled changes for each participant are illustrated in *Figure B1*. Mean heart rate for each condition and mean activity counts are illustrated in *Figures B2 and B3.* Overall, median temper outburst behaviours across all participants were similar in no change, un-signalled and signalled change conditions (no change: 0.78; un-signalled change: 0.81; signalled change: 0.68). Mean heart rates and activity counts were also similar (*heart rate*: no change: 75.61; un-signalled change: 74.04; signalled change: 74.28; *activity count:* no change: 74.57; un-signalled change: 66.47; signalled change: 73.04).

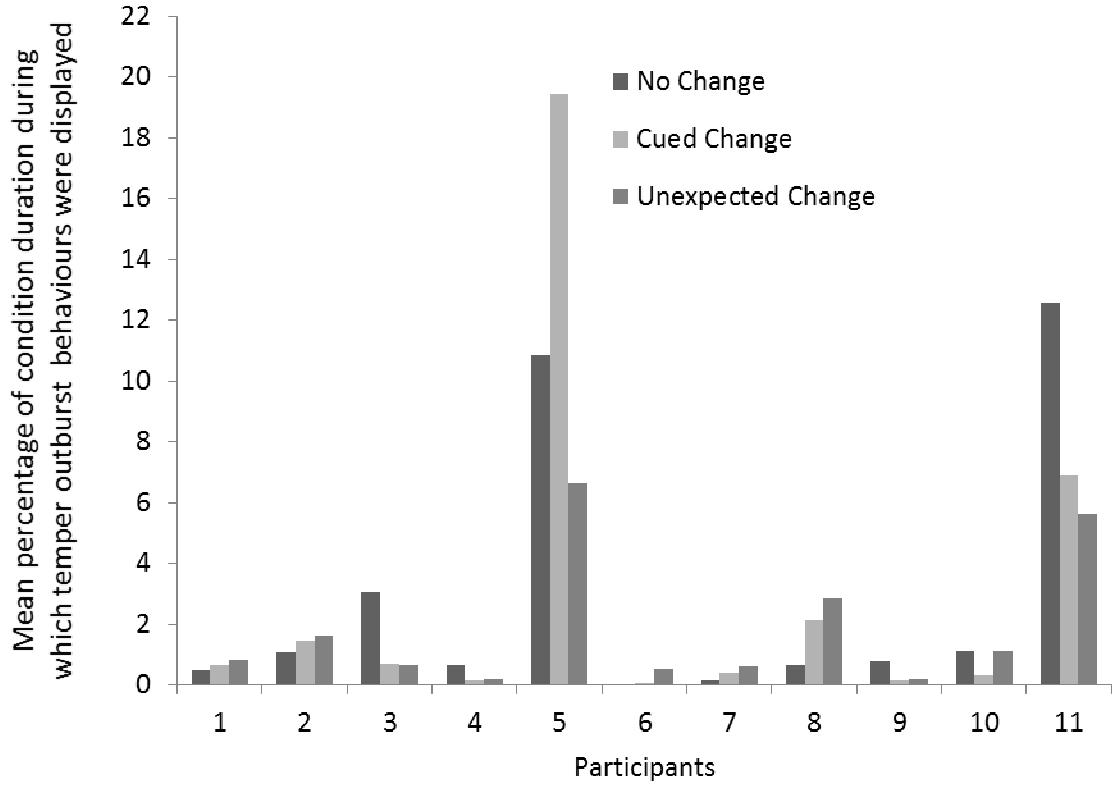


Figure B1. Temper outburst behaviours demonstrated during test conditions in which no changes were imposed (no change), signalled changes (cued change), or un-signalled changes (unexpected change) were imposed.

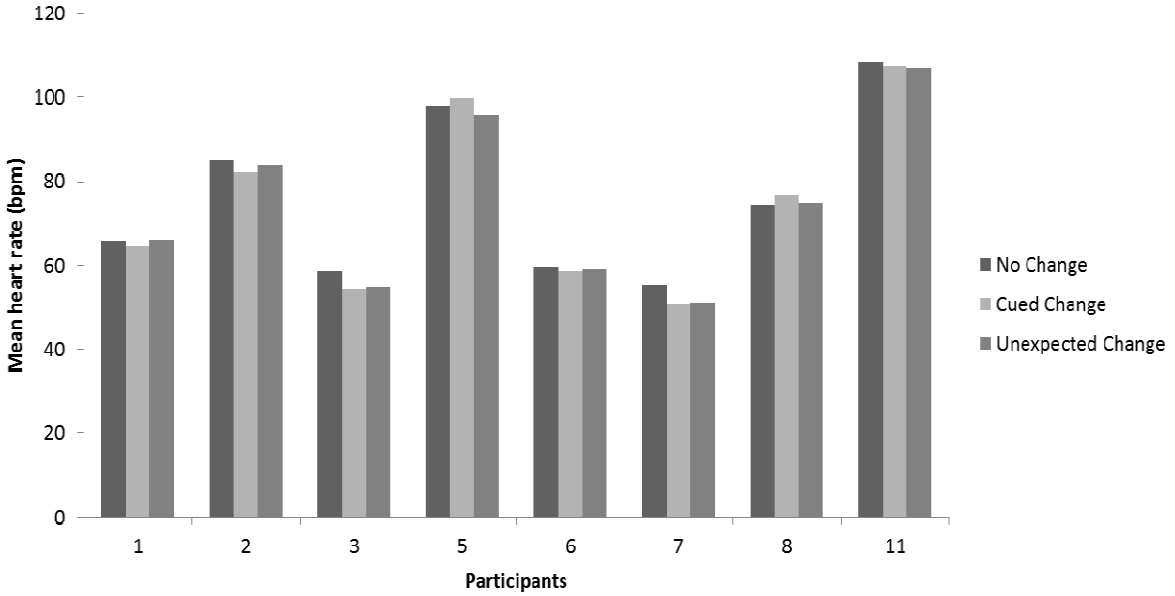


Figure B2. Heart rate demonstrated during test conditions in which no changes were imposed (no change), signalled changes (cued change), or un-signalled changes (unexpected change) were imposed.

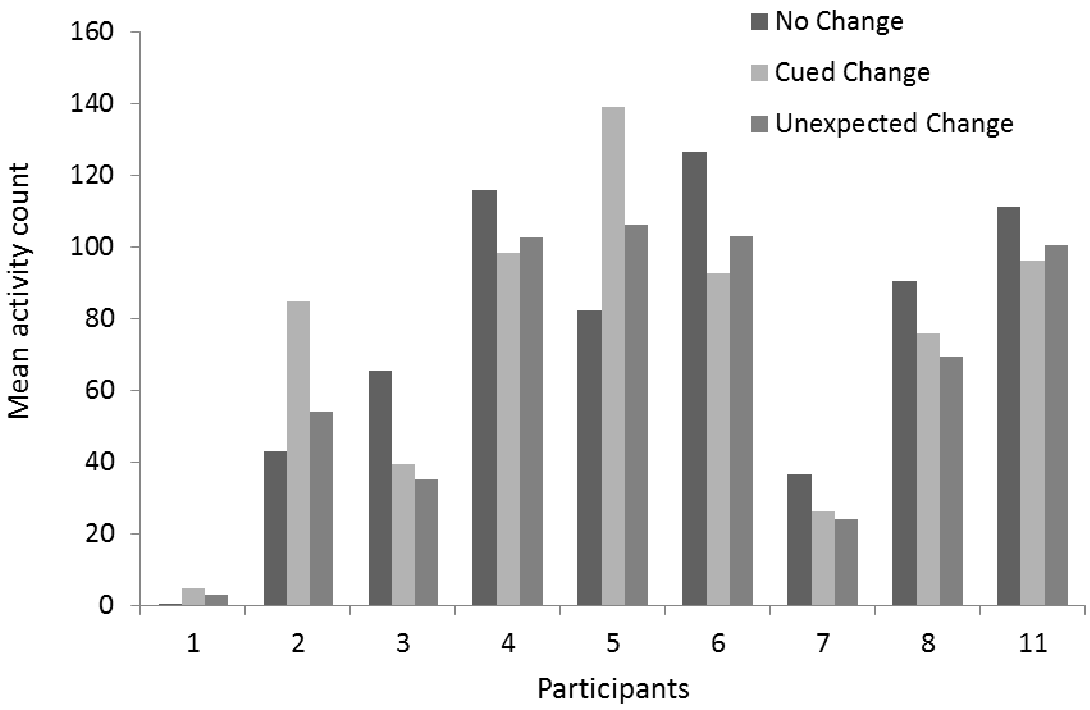


Figure B3. Activity count demonstrated during test conditions in which no changes were imposed (no change), signalled changes (cued change), or un-signalled changes (unexpected change) were imposed

Appendix C. **Written information sheet provided to caregivers**

Background

The signal we have given to you should be used to help you to signal any change to routine/expectation that happens to <participant>. The signal should be something new and distinctive for <participant>. We will help to teach <participant> to understand what the signal means.

We hope that with enough practise <participant> will learn that the signal means that something **will** change. We think that this will make it easier for <participant> to deal with the change because <he/she> will be expecting it to happen so <he/she>will be prepared.

We/you will not know in advance about all changes that will happen so we cannot signal all changes. What is important though is that **every time** <participant> sees the signal a change **must** happen. You should **only** show the signal when you know that a change to routine or expectation is going to happen.

How to use it?

Anytime that you become aware of a change to <participant's> routine or expectation you should be prepared to use the signal. Where possible the signal should be kept with you. In the usual way that you might address a change with <participant> do the same as you normally would but always pair this with the signal and the phrase "This picture means that something different is going to happen. (Then explain change as you normally would)."

So:

1) If you would usually talk about the change a few times before the event show the signal every time you talk to <participant> about it.
2) If you usually only address the change once show the signal this one time.
3) If you find out just before a change that it is about to happen show the signal as soon as you can.
4) If a change happens and you did not find out about it in advance do not worry. It might not be possible for you to signal every change but what is important is that you never show <participant> the signal unless a change is going to happen.

Important rules:

1. Not every change has to be cued but it is important that you should **only** show the signal if a change is going to happen. **Do not** show the signal if a change to routine/expectation is not going to happen.

2. When using the signal always use the phrase "This picture means that something different is going to happen. (Then explain change as you normally would)."

3. Always think about changes to <participant's> routine or <participant's> expectations. It may be that sometimes a change happens that you do not think <participant> would be aware of. If this happens do not signal the change. You

should only signal changes that you think are changes from <participant's> point of view.

4. If a change is signalled and <participant> does not display a temper outburst then tell <participant> that they have dealt with the situation very well and praise <him/her> for this achievement.

What you need to do over the next 4 weeks

Week 1:

- Carry out the strategy outlined above but by only cueing those changes that are less problematic that we have identified in the hierarchy.
- To help <participant's>learning try to add a small change to <his/her> routine/expectation at least once every two days and make sure you use the signal to warn <participant> of this change. This will help <participant> to remember the purpose of the signal. The changes do not need to be big. Changes can be made where you would not expect <participant> to find it very difficult.
- Continue to use the behaviour diary –
    - Record all changes to routines or expectations that occur that you are aware of even if there is no outburst. Times when there is no outburst but there is a change write down the time and date and tick either the change to routine or change to expectation box in the "Before" column and leave everything else blank.
    - Please document for all times that change occurs (both when there is an outburst and when there is not an outburst) briefly what the change was.

Week 2:

- Carry out the strategy outlined above but now cue all changes that occur and all changes that we identified in the hierarchy.
- To help <participant's>learning try to add a small change to <his/her> routine/expectation at least once every two days and make sure you use the signal to warn <participant> of this change. This will help <participant> to remember the purpose of the signal. The changes do not need to be big. Changes can be made where you would not expect <participant> to find it very difficult.
- Continue to use the behaviour diary –
    - Record all changes to routines or expectations that occur that you are aware of even if there is no outburst. Times when there is no outburst but there is a change write down the time and date and tick either the change to routine or change to expectation box in the "Before" column and leave everything else blank.
    - Please document for all times that change occurs (both when there is an outburst and when there is not an outburst) briefly what the change was.

For the next 2 weeks:

- Carry out the strategy outlined above

- This time continue as you would normally. There is no need now to deliberately make any changes. Just signal any changes that happen anyway.
- Continue to use the behaviour diary –
    - Record all changes to routines or expectations that occur that you are aware of even if there is no outburst. Times when there is no outburst but there is a change write down the time and date and tick either the change to routine or change to expectation box in the "Before" column and leave everything else blank.
    - Please document for all times that change occurs (both when there is an outburst and when there is not an outburst) briefly what the change was.

If you need help or advice

Contact me on [telephone numbers] or [email address]
We will contact you on a weekly basis to check if everything is going ok

Appendix D

Table D1 Operational definitions and Kappa inter-rater reliability coefficients for temper outburst behaviours observed during the *Mechanistic Evaluation Sub-Procedure*.

Behaviour	Operational Definition	Kappa
Questioning	The participant asks their parent/carer/teacher researcher a question.	0.62
Ignoring Requests	Participant does not respond to a verbal request made by their caregiver or verbalises about something unrelated to the request. This should be coded until a further verbal response from the caregiver (a further request or a verbalisation about something unrelated to the request) or the participant initiates a response.	0.95
Arguing	Participant makes verbalisations in the form of statements of disagreement, giving orders or making demands.	0.92
Crying	Participant shows tears or speech or non-speech vocalisations associated with crying.	0.96
Verbal Aggression	Participant verbalises threats or makes hurtful comments towards their caregiver. This could also include any offensive language.	0.87
Picking Nose	Coded for Matthew only. Participant picks nose with fingers or tissue. Includes blowing nose and eating any mucus from fingers or tissue.	1.0

***Appendix E*. Example of a behaviour diary entry sheet**

Date	Time	Behaviour Observed	Duration	Intensity 1-10	Before	After
		€ Shouted € Kicked € Spat at someone € Red Face € Stamped feet € Other_____			€ Change to routine € Change to expectation € Somebody told him off € Somebody told him off about food € Did not get some food he wanted € Other_____	€ Cried € Said sorry repeatedly € Blamed somebody else € Wanted a cuddle € Tired € Other _____

Appendix F. Supplementary results

Behaviour diary reports were examined over the four week baseline period to check for trends that would imply temper outbursts were increasing or decreasing during that period (*Table F1*). No consistent trends were observed. Nevertheless, the weekly records of temper outbursts clearly indicate substantial variability from week to week across all participants.

Table F1. Number of change triggered temper outbursts demonstrated in each week of the four week baseline period for each participant

Participant	Week 1	Week 2	Week 3	Week 4
Daniel	3	3	1	0
Alfred	1	2	0	0
Charles	1	1	1	0
Scott	1	0	0	0
Bob	0	0	1	0
Matthew	2	0	0	2
John	0	0	0	0
Peter	0	0	0	1
James	0	0	0	2

Anecdotal open ended reports to inform on the process of administering the signalling intervention were ascertained from caregivers on a weekly basis via telephone. Caregivers' responses are summarised in *Table F2.*

Table F2 Caregivers' responses to weekly open ended reports on the process of administering the signalling intervention

Participant	**Anecdotal information provided on use of the signal in the participant's natural environment**
Alfred (Parent)	He is into it in a big way, he loves the card
	He asks for the card when he thinks there might be a change
	He has been better behaved, I'm grateful of the quiet time
	He knows what the card means
	Said he likes to see the card
	With the card he accepts these changes
Charles (Parent)	The card quickly became a no-no
	We have used the word 'change' as a means of approaching the problem and as a strategy appears to be working
Scott (Parent)	It does not always work
	....is going ok, although I need a card tattooed to my hand
Bob (Care home)	Don't need card now, just say 'change'
	Recently had to make a change to food, the hardest change for..., he just accepted it
	Good idea, Is working
	He likes it and says it's useful
	Managed to cut the number of cigarettes down a day by using the card and approaching it as a change
Matthew (Participant)	It is helping
John (School)	....has learned to identify the card
	....this has reduced minor challenging behaviour, especially if the change is not to do with food
	This has been especially helpful with changes in his timetable
	It has even worked with some bigger changes
	It will take a long time and constant work for....to accept changes to his food routine
Peter (Care home)	The cards are positive
	Planned changes before the stress builds up can be helped with cards
	If....is juggling too many....worries....then the card is less effective
James (Parent)	He has been a lot calmer

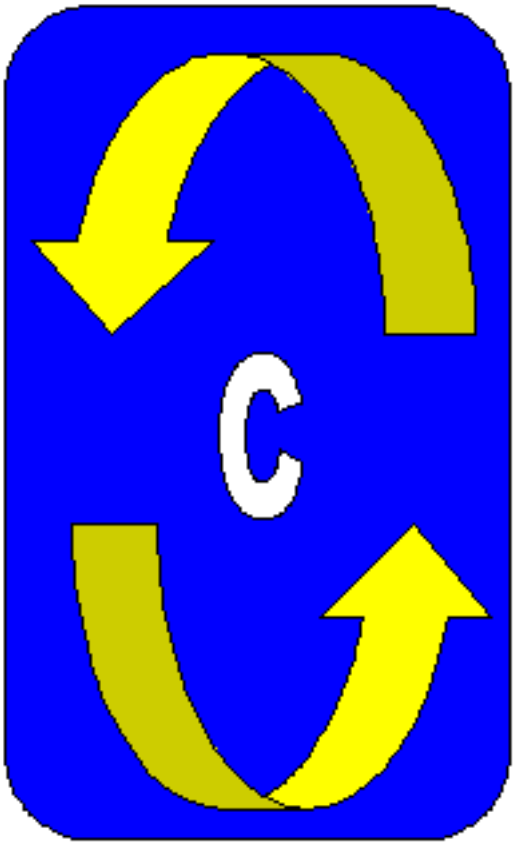


Figure 2

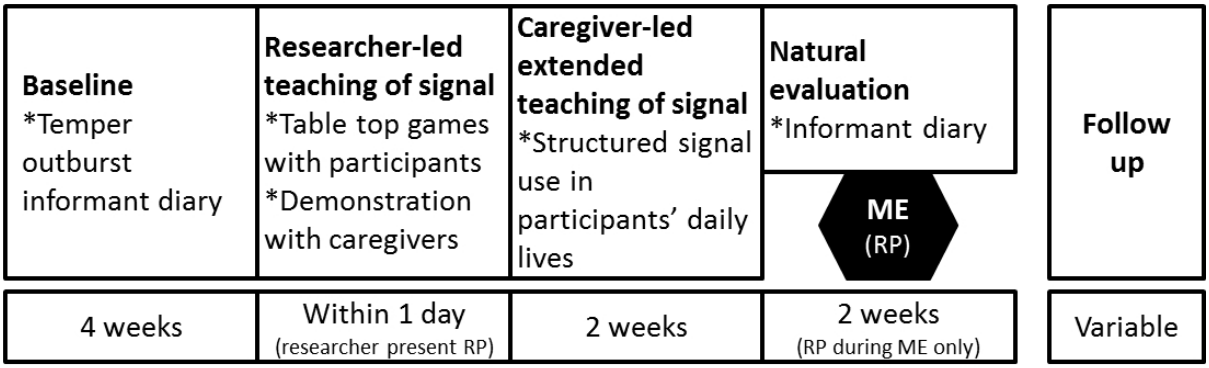


Figure 3

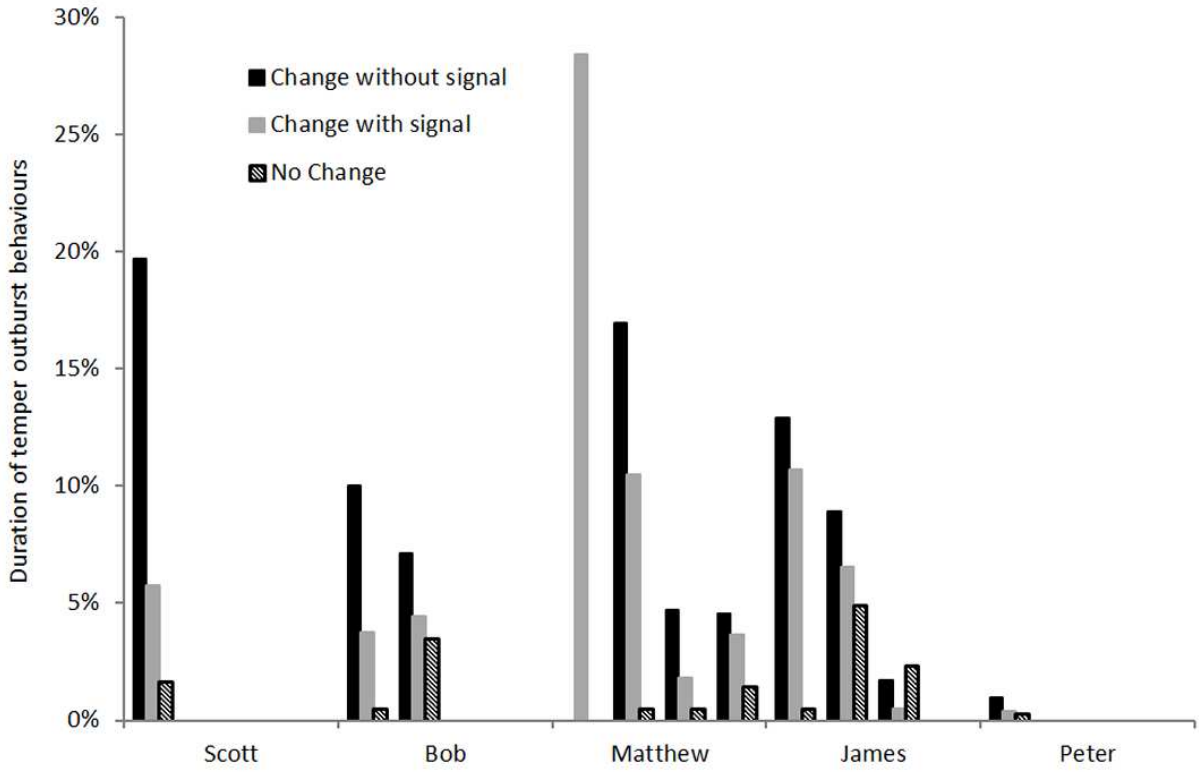
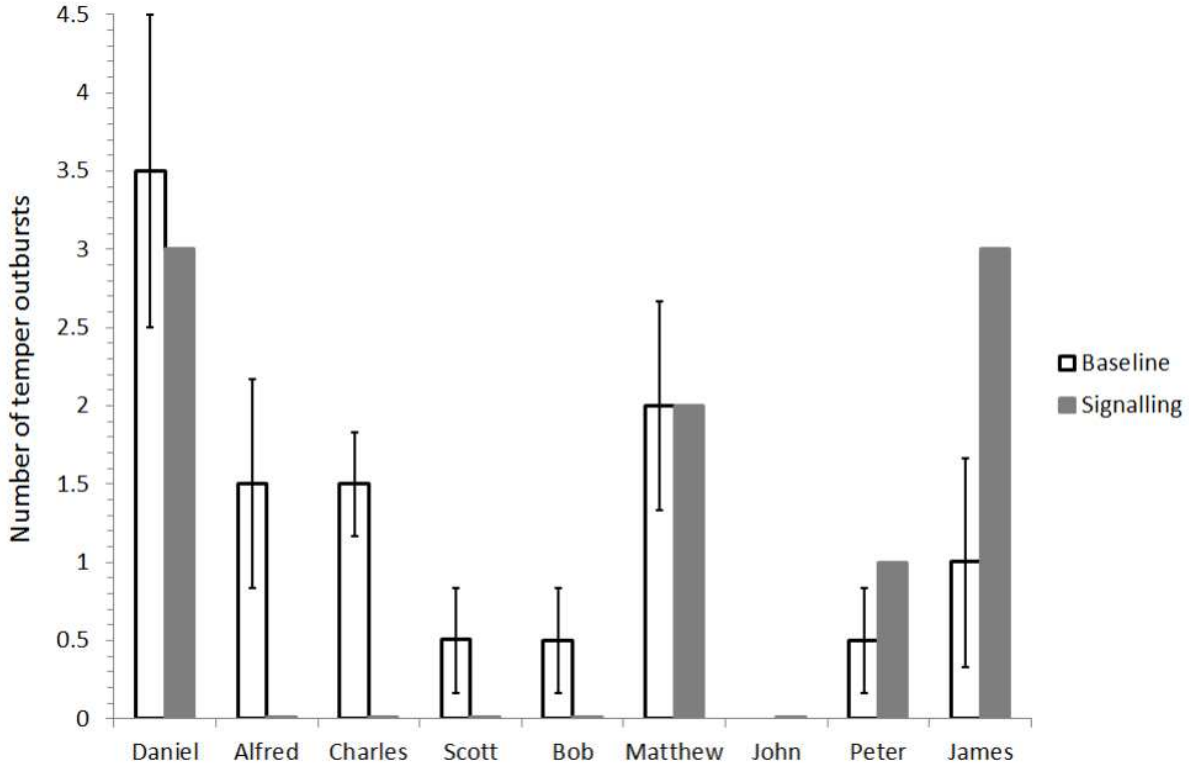


Figure 4



- Caregivers provided a warning signal before a forthcoming change to participants' expectations.
- Participants' showed less challenging behaviour following signalled than not signalled changes.
- Diaries showed some reliable reductions in temper outbursts during signalling use.
- Caregivers appear willing and able to use the signal.